

#### CLINICAL PRACTICE

# Tic Phenomenology and Tic Awareness in Adults With Autism

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ABSTRACT: Background: Tics are common in people with autism spectrum disorder (ASD). However, their phenomenology and characteristics have not been studied in detail. Methods: Based on video sequences of 21 adults with ASD without intellectual disability and 16 adults with Gilles de la Tourette syndrome (GTS), tic severity, tic repertoires, and tic awareness were determined. Results: Ten ASD and all GTS participants had tics during video recordings. The ASD group had significantly fewer tics, compared to GTS. Tic distribution and tic repertoires were comparable, but more restricted in ASD. All GTS participants, but only 5 of the 10 ASD participants, were aware of their tics. Conclusions: Tics are common in adults with ASD. They are indistinguishable from tics in GTS and are similarly distributed, but less severe. Tic awareness is limited in ASD.

Tics resemble patterned normal movements, but appear repetitively, often with exaggerated intensity and increased frequency. Although their neurophysiological properties are largely indistinguishable from voluntary movements, 1,2 tics appear without appropriate context as rather uncontrollable and intrusive fragments of motor behavior. Tics can occur in different conditions. 3 However, the vast majority of tics can be classified as primary tic disorder, with Gilles de la Tourette syndrome (GTS) being the most relevant. Most knowledge on tic phenomenology stems from research in these patients.

GTS is a neurodevelopmental disorder with a mean age of onset around 7 years of age and prevalence between 7 and 10 per 1,000 at that age. 4.5 Tics in GTS fluctuate over time in intensity, frequency, and characteristics and are typically preceded by premonitory sensations or urges. Associated motor phenomena, conceptualized under the rubric of complex tics, include coprophenomena (obscene and socially inappropriate, often offensive gestures or utterances) and echophenomena (imitation of actions or sounds without explicit awareness). Commonly associated psychiatric comorbidities are attention deficit hyperactivity disorder (ADHD) and obsessive compulsive behavior/disorder (OCB/D). Moreover, there is increasing evidence that autism spectrum disorder (ASD), another common neurodevelopmental disorder, is also over-represented in

the GTS population. For example, Freeman et al. reported a prevalence of 4.5% of ASD in a large multisite cohort of 3,500 patients with GTS,<sup>6</sup> which was later replicated by the same group in a larger sample of 7,288 patients.<sup>7</sup> As a corollary, tics are also common in ASD. Current prevalence estimates range between 22%<sup>8</sup> and 34%,<sup>9</sup> depending on the studied population and applied screening tools. These data and the recognition of additional phenomenological overlap between these neurodevelopmental disorders, including presence of echophenomena,<sup>10</sup> sensory hypersensitivity to exteroceptive stimuli,<sup>11,12</sup> and the common occurrence of ADHD and OCD in both disorders, have fueled interest in the search of shared genetic backgrounds and elucidation of common pathophysiological mechanisms.<sup>13,14</sup>

However, although widely recognized, only scarce information exists on the exact phenomenological characteristics of tics in ASD.<sup>8,9,15–17</sup> This can be explained by clinical heterogeneity of ASD and methodological difficulties in assessing different co-occurring hyperkinetic phenomena in ASD. Stereotypies, for example, are common repetitive motor behaviors that are part of the diagnostic criteria of ASD.<sup>18</sup> Although they are distinct from tics, they can be mislabeled as such.<sup>17,19</sup> Criteria to differentiate tics and stereotypies on clinical grounds have been proposed, <sup>17,20</sup> but methodological differences related to recognition and classifi-

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cation of extra movements in ASD have hampered tic characterization.

Therefore, the aim of this study was to explore similarities and differences in tic phenomenology and awareness in a well-characterized sample of adults with ASD without intellectual disability and to compare them to patients with uncomplicated ("pure") GTS. The modified rush video-based tic rating scale (MRVS<sup>21</sup>), a valid instrument for objective clinical evaluation of tics, was employed.

### **Methods**

### **Participants**

ASD and GTS participants were assessed in the outpatient clinic in the Departments of Neurology and Psychiatry at the University Medical Center Hamburg. In order to ensure study compliance and accurate self-reports, 21 adult ASD participants without intellectual disability (IQ ≥70) were consecutively recruited. All had been diagnosed by independent clinicians before the study and were additionally rediagnosed by a psychiatrist (D.S.) as either having autistic disorder or Asperger's (F84.0 and F84.5) during a psychiatric interview using established criteria for ASD according to the International Classification of Diseases, Tenth Revision.<sup>22</sup> D.S. also assessed the Structured Clinical Interview for Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV), axis 1 and 2 disorders. 23,24 ASD participants were characterized by an IQ >70 (mean = 112.95; range = 92.0-143.0; standard deviation [SD] =  $\pm 15.64$ ) in tests for verbal intelligence (German Multiple Choice Word Test [IQ-MWT-B]), 25 hence labeled as "without intellectual disability." Autistic trait severity was assessed using the Autism Spectrum Quotient (AQ),26 Systemizing Quotient (SQ),<sup>27</sup> Empathy Quotient (EQ),<sup>28</sup> and the "Reading the Mind in the Eyes" Test (Eyes Test). 29 None of the ASD participants had been previously examined for the presence of tics.

GTS participants were diagnosed by neurologists specialized in movement disorders (A.M., C.G.), using DSM-IV, Text Revision (DSM-IV-TR), criteria.<sup>30</sup> Only individuals with uncomplicated GTS (i.e., without ADHD or OCB/D; methods previously described<sup>31</sup>) were included in the study because we were predominantly interested in the group comparison of tics and not other extra movements that may occur in ADHD or OCB/D (e.g., impulsive hyperkinesias in the former and compulsions in the latter).

All participants gave written informed consent before study attendance. The study was performed in accord with the Declaration of Helsinki and its later amendments and was approved by the local ethics committee.

### Tic Analysis

Participants were filmed in a standardized setting. Tics in both groups were evaluated three times as follows: First, video sequences were evaluated according to the MRVS protocol.<sup>21</sup> All participants were videotaped sitting on a chair alone in the room while not suppressing their tics ("free ticcing"). A close-up

2.5-minute sequence of the head and shoulders was filmed, followed by a whole-body sequence of equal duration. MRVS total score and subscores (motor tic frequency, motor tic severity, number of affected body areas, phonic tic frequency, and phonic tic severity) were determined. Second, the same video recordings were re-evaluated for motor tic distribution in five body parts (head, neck/shoulders, arms/hands, legs/feet, and trunk). Two different analyses were employed: (1) number of patients with tics in a given body part and (2) pooled number of tics per body part across participants expressed as a percentage of the total number of tics. Finally, tic repertoires were assessed by characterizing each subject's individual motor tics during video recordings. During the evaluation of video sequences of ASD participants, stereotypies were determined according to proposed criteria 17,20 and excluded from further analysis (list of stereotypies is provided in Supporting Table 1). Video analyses were performed by a movement disorders specialist (C.G.).

In addition, participants were offered a definition of tics and were then asked whether they were aware of their presence.

Mann-Whitney U test and Student's t tests for independent samples were used to calculate between-group differences. Significance level was set at P < 0.05.

### Results

Group characteristics are given in Table 1.

All GTS participants and 10 of 21 ASD participants had motor tics during the assessed periods. Total MRVS scores were significantly lower in the ASD group with tics, compared to GTS, owing to significantly lower MRVS subscores for motor tic frequency, severity, and number of affected body parts (see Table 2).

Assessments of motor tic distribution showed comparable results in both groups (GTS and ASD with tics) following a rostrocaudal gradient. However, motor tic distribution was restricted in ASD (Fig. 1A,B).

The motor tic repertoire of ASD participants with tics was similar to that of GTS participants (see Fig. 2). However, participants with GTS displayed "orchestrated" tics (multiple tics at the same time, or continuous ticcing; 48 different tics; Supporting Table 2) more often than ASD participants (16 different tics; Fig. 2). This was also reflected by higher MRVS subscores for motor tic severity in GTS (2.7  $\pm$  1.14), compared to ASD (1.1  $\pm$  0.32; U = 18.50, z = -3.445, P = 0.001; see Table 2).

All 16 GTS participants reported to be aware of having tics. In ASD, only 5 of the 10 participants with tics were aware of their presence. ASD participants with tic awareness scored lower on AQ and SQ, but higher on EQ and Eyes Test than ASD participants who were not aware of having tics (Table 3).

### **Discussion**

Tics were common in our studied ASD sample. Tic distribution and repertoires were similar to GTS, but they appeared to be less severe and less frequent. Awareness of tics was limited in ASD.

**TABLE 1** Participants' characteristics

Characteristic	ASD (n = 21)	GTS (n = 16)	
Sex (male/female)	12/9	15/1	
Mean age in years $(\pm SD)^a$	$34.6 \ (\pm 8.48)$	29.9 (±8.45)	
Verbal intelligence quotient MWT-B (±SD)	113.0 (±15.64)	NA	
Comorbid psychiatric disorder	11	0	
	[6× recurrent depressive episodes only;		
	1× bipolar disorder only;		
	2× recurrent depressive episodes and ADHD;		
	1× recurrent depressive episodes and OCD;		
	1x depression and panic disorder]		
Psychopharmacological medication	7	4	
	[3× SSRI;	[2× tiapride;	
	3 SNRIs;	1× aripiprazole	
	1 promethazine]	1× levodopa <sup>b</sup> ]	

Clinical characteristics of ASD and GTS participants. n, total number of subjects.  $^{\rm a}P=0.1$ .

<sup>b</sup>For Restless Legs Syndrome.

NA, not available; SSŔI, selective serotonin reuptake inhibitor; SNRI, serotonin-norepinephrine reuptake inhibitor.

TABLE 2 Modified rush video-based scale tic ratings

Characteristic	Scale	ASD (n = 21)	GTS (n = 16)	P Value
Percentage of participants having motor tics		47.6	100	
Percentage of participants having phonic tics		9.5	37.5	
Motor tic frequency: tics/min (±SD)		5.4 (±2.8)	25.5 (±19.3)	<0.001 <sup>a</sup>
Phonic tic frequency: tics/min (±SD)		0.4 (±0.8)	1.2 (±2.1)	0.452
MRVS subscore: number of body areas (±SD)	0–4	1.1 (±0.3)	3.0 (±0.8)	<0.001 <sup>a</sup>
MRVS subscore: motor tic frequency (±SD)	0–4	1.0 (±0.0)	1.8 (±0.9)	0.017 <sup>a</sup>
MRVS subscore: phonic tic frequency (±SD)	0–4	0.2 (±0.4)	0.5 (±0.7)	0.421
MRVS subscore: severity of motor tics (±SD)	0–4	1.1 (±0.3)	2.7 (±1.1)	0.001 <sup>a</sup>
MRVS subscore: severity of phonic tics (±SD)	0–4	0.2 (±0.4)	0.5 (±0.8)	0.452
MRVS total score (±SD)	0–20	3.6 (±1.4)	8.4 (±3.1)	<0.001 <sup>a</sup>

<sup>&</sup>lt;sup>a</sup>Level of significance at 0.05 (Mann-Whitney's U test). n, number of participants.

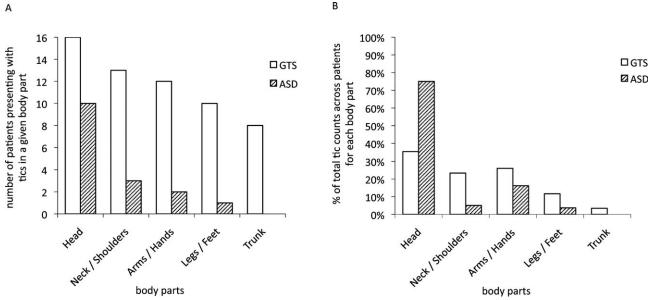


Figure 1 Motor tic distribution. (A) Number of patients with tics in a given body part. (B) Pooled number of tics per body part across participants expressed as a percentage of the total number of tics (100%) for each group (16 GTS and 10 ASD participants). GTS, white bars; ASD, dashed bars.

In keeping with previous reports, 8,9 ASD participants with tics were less affected than GTS patients, reflected in overall lower MRVS total scores and subscores. For example, in the large pediatric ASD sample of Baron-Cohen et al., tic severity evaluations ranged in the lower third of the Yale Global Tic Severity Scale. Similarly, Canitano and Vivanti reported comparable findings and further showed that tic severity was associated with levels of cognitive impairment.8 Children with

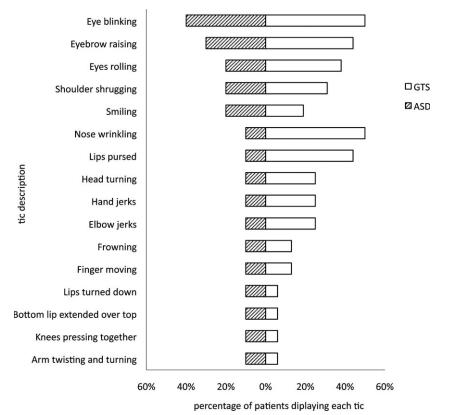


Figure 2 List of individually occurring motor tics of ASD (n = 10, dashed bars) and GTS (n = 16, white bars) participants and their prevalence. n, number of patients. [Correction added on 6 April 2015, after first online publication: opening sentence of figure caption removed]

TABLE 3 Tic awareness and autistic trait severity

Characteristic	ASD With Tic Awareness (n = 5)	ASD Without Tic Awareness (n = 5)
AQ (±SD) SQ (±SD) EQ (±SD) Eyes Test (±SD)	35.2 (±4.6) 26.4 (±9.6) 16.2 (±4.3) 22.0 (±3.9)	41.6 (±4.4) 40.6 (±15.8) 13.6 (±9.5) 19.2 (±7.0)

n, number of participants.

milder developmental impairment also had fewer tics. The results of our sample of adults with ASD without intellectual disability further corroborate this finding. In addition, for both GTS and ASD, a similar pattern of rostrocaudal tic distribution was found, with the head invariably being affected in all participants with tics. Tic repertoires were also similar in adults with ASD and GTS, but more restricted in the former, possibly reflecting overall lower tic severity.

The prevailing model of tic pathophysiology in GTS suggests excess motor generation as a result of disinhibition at somatotopically arranged cortical and subcortical levels, including the basal ganglia. 32,33 Our data show, for the first time, that the somatotopical gradient of tic-related motor disinhibition is also present in adults with ASD. This may suggest a common neural basis of tics in both disorders. Although no studies have addressed tic pathophysiology in ASD to date, the basal ganglia and related corticostriatal circuits have also been

related to the abnormal repetitive motor output of stereotypies in ASD.  $^{34-36}$ 

Only half of the 10 ASD participants with tics reported being aware of them. The absence of tic awareness in some ASD adults may reflect a primary deficit of self-referential cognitive processing. 37,38 However, it might also reflect a secondary effect related to the altered perception of ASD adults of their social environment. For example, children with ASD have difficulties in recognizing self-conscious emotions, such as shame and embarrassment, as a consequence of lack of understanding social norm violations and negative social evaluations.<sup>39</sup> In GTS, it has been proposed that awareness of tics might gradually develop over time as a response to children's growing sensitivity, and therefore selfawareness, to negative social reactions related to excessive involuntary movements. 40 Individuals with ASD may be less prone in perceiving social distress as response to their motor behavior and may therefore consequently fail to become more aware of their tics. This notion is supported by the fact that participants who were not aware of their tics had lower scores on EQ and Eyes Test paralleled by higher scores on AQ and SQ. These tests are used to assess the ability of empathy (EQ)<sup>28</sup> and emotion recognition in others (Eyes Test)<sup>29</sup> and serve as indicators of autistic trait severity (AQ and SQ).<sup>26,27</sup>

This study has several limitations. The patient collective consisted of ASD participants without intellectual disability, as well as uncomplicated ("pure") GTS adults, and is thus not

representative of the entire ASD and GTS spectrum. Also, the examined sample size is relatively small, compared to previous studies, and results can therefore not be generalized. However, the strength of this study lies in the inclusion of a relatively homogenous, well-characterized group of adults with ASD. Indeed, the absence of intellectual disability of our ASD sample facilitated precise understanding of tests and questionnaires, as well as excellent study compliance. On the other hand, the investigation of uncomplicated ("pure") GTS participants reduced the influence of comorbidities and allowed for reliable inferences as to the core of tic phenomenology. It should, however, be noted that the MRVS might not be best suited for evaluating tics in patient samples other than GTS, given that tics appear less frequently in ASD and may be missed during the relatively short period of evaluation. On the other hand, the MRVS is the most widely used validated and reliable<sup>21</sup> videobased tic evaluation method available. Finally, we cannot exclude the possibility that the overall mild tic severity of studied adults with ASD might have contributed to limited tic

To conclude, although tics in adults with ASD seem to be milder in severity and frequency than in GTS patients, a rostro-caudal body tic distribution gradient is characteristic for both disorders. Adults with ASD appear to have reduced tic awareness. This indicates that, at least in some cases, tics may occur independent of attentional focus and without explicit awareness, which, in turn, might be further influenced by understanding of social norms.

### **Author Roles**

(1) Research Project: A. Conception, B. Organization, C. Execution; (2) Statistical Analysis: A. Design, B. Execution, C. Review and Critique; (3) Manuscript Preparation: A. Writing of the First Draft, B. Review and Critique.

U.K.: 1B, 1C, 2B, 3A

O.S.: 1B, 1C, 2B, 3B

D.S.: 1C, 3B

N.D.: 1B

V.B.: 2B, 2C, 3C

T.B.: 2C, 3B

V.R.: 3B

A.M.: 1A, 2C, 3B

C.G.: 1A, 1B, 1C, 2A, 2B, 2C, 3A, 3B

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## **Supporting Information**

Additional Supporting Information may be found in the online version of this article:

**Table S1.** List and prevalence of stereotypies of adults with autism spectrum disorder captured during video recordings

**Table S2.** List and prevalence of individually occurring motor tics (i.e., tic repertoire) of GTS (n = 16) and ASD (n = 10) participants.